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**Article** *in* JPO Journal of Prosthetics and Orthotics · April 2021

DOI: 10.1097/JPO.0000000000000358

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# Patient-Reported Efficacy of the University of California, San Francisco, Custom Pectus Carinatum Orthosis

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## ABSTRACT

**Introduction:** Pectus carinatum is a bony deformity of the anterior chest wall and has a strong negative impact on patient self-esteem and quality of life. During adolescence, the deformity is often flexible and can be treated with a compressive orthosis. Compliance with orthoses yields deformity correction and self-esteem improvement (*J Pediatr Surg*. 2013;48:1055-1059). The purpose of this study is to evaluate the compliance to and patient satisfaction with the custom University of California, San Francisco (UCSF), Pectus Carinatum Orthosis (PCO) design.

**Materials and Methods:** This was a cross-sectional survey of the patients who received orthotic treatment for pectus carinatum at UCSF between August 2012 and June 2018. Potential subjects were contacted and asked to complete the Pectus Carinatum Evaluation Questionnaire (PCEQ), which was administered online via Research Electronic Data Capture. The PCEQ measures compliance as well as the physical and psychosocial impact of orthotic treatment for pectus carinatum. Results were compiled and summarized using nonparametric descriptive statistics.

**Results:** Of the 35 consented subjects, 12 (11 male patients and 1 female patient aged 12–17 years) completed the survey. Subjects reported an average wear time of 12.7 hours per day, 5 days a week. Seven reported no symptoms, three experienced chest pain, two reported difficulty breathing, and one had back pain. Eight reported happiness with the results of their orthotic treatment.

**Conclusion:** Wear times reported by the subjects were comparable with those reported in other studies looking at prefabricated pectus orthoses. However, there were lower reports of pain associated with the UCSF treatment than in other studies. Further research is necessary to determine the benefit of custom fabricated versus prefabricated PCOs. (*J Prosthet Orthot*. 2021;33:96–100)

KEY INDEXING TERMS: pectus carinatum, chest wall deformity, compressive orthosis

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Pectus carinatum (PC), also known as "pigeon chest," is the second most common anterior chest wall deformity in the United States,[1,2] with an estimated prevalence of 0.6%.[3,4] It is characterized by a protrusion of the sternum most commonly attributed to costal cartilage overgrowth. It affects males more than females and is diagnosed most often in adolescence during periods of rapid growth. Family history is noted in 25% to 33% of cases,[2,5] but there are no currently identified genetic indicators. The deformity rarely results in cardiopulmonary or respiratory symptoms but can have a strong negative impact on a patient's self-esteem and quality of life.[1,2,5] Multiple studies have found that individuals with PC will avoid activities such as swimming where it is more likely that their deformity will be noticed.[1,4,5] Body image is so impacted that Steinmann et al.[1] found that 94.7% of patients seeking corrective surgery cited cosmesis as one justification for the procedure, and 68.4% sought surgical correction solely because of cosmesis.

Surgery, specifically the modified Ravitch procedure,[4,6] has been the accepted treatment for PC for the past 50 years. This procedure involves resection of costal cartilage overgrowth and a sternal osteotomy. Overall, surgery yields positive outcomes for patient quality of life, mental health, and self-esteem,[2,7] although it subjects the patient to the risks of general anesthesia as well as postoperative pain and recovery.

More recently, anterior compression orthotic intervention has been viewed as a viable nonoperative treatment for PC. In the early 1970s, torso casting was performed in an attempt to correct the deformity, but it was met with minimal success.[5] Case reports on orthosis use in the 1990s had more promising results.[5,6,8,9] The literature shows that compliance with orthoses yields deformity correction,[3,4,6,8,10] minimal recurrence,[6,8] and significantly improved self-esteem.[3,4]

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Supplemental digital content is available for this article. Direct URL citations appear in the printed text and are provided in the HTML and PDF version of this article on the journal's Web site (https://journals.lww.com/jpojournal/pages/default.aspx).

Disclosure: The authors declare no conflict of interest.

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The biomechanical concept for PC orthoses is based on Wolff's law,[11] which states that bone and cartilage gradually remodel under pressure. Constant anterior-posterior compression of the prominence in a flexible deformity therefore should encourage more normal development of the costal cartilage and sternum. Like scoliosis, flexibility of the deformity tends to decrease as growth plates fuse and the patient's Risser sign increases.[11] The Calgary protocol[4,12] is most commonly used, although there is little research comparing its efficacy to alternative wear schedules. It entails 23 hours of wear per day, 7 days per week until correction is achieved. At that point, the patients enter a maintenance phase during which they wear the orthosis 8 hours per day (typically at night) until skeletal maturity to prevent reoccurrence. This transition to nighttime wear is generally considered a marker of successful orthotic intervention, although currently, there is no standard for acceptable degree of correction. At University of California, San Francisco (UCSF), the standard of care is reduction to less than 1/8 inches of asymmetry, at which point the deformity is mild and the orthotist will begin discussing transition to nighttime wear if the patient is satisfied with his or her chest appearance. Compliance with the Calgary wear protocol is the factor most highly correlated with the success of the orthotic treatment.[3,5,6,9,13]

Prefabricated pectus orthoses generally consist of contourable anterior and posterior aluminum bands with pads and ratchet straps on both sides. With this design, some common complaints include discomfort, bulk, migration, skin irritation, and breathing restriction. In addition, they are symmetric in design, whereas most cases of PC are asymmetric. The UCSF Pectus Carinatum Orthosis (PCO) (Figure 1a, b) is a custom-fabricated orthosis designed to more intimately contour around the patient's anatomy. It uses a 6-mm mineral oil gel pad (Pedifix Inc, Brewster, NY, USA) over the apex of the prominence to distribute pressure across the prominence and improve comfort. Velcro straps are used rather than ratchets for reduced visibility and ease of replacement. The orthosis has clearance for the pectoralis muscles and allows for full lateral expansion of the chest cavity. Currently, there is no literature on this specific design and the physical or psychosocial impact.

The purpose of this retrospective study is to describe self-reported outcomes of patients who used the custom UCSF-PCO. It is hypothesized that subjects will report compliance with prescribed wear time, that at least 80% of patients polled will report satisfaction with orthosis experience, and that patients who received the UCSF-PCO will report an overall positive experience with minimal physical or psychosocial impact.

## MATERIALS AND METHODS

### SUBJECTS

Pediatric patients who sought orthotic treatment for PC at UCSF between August 2012 and June 2018 were recruited for this study. They were contacted via phone. Subjects were excluded if they were over older than 18 years, if they could not understand provided instruction, if they were a ward of the state, or if they or their guardians were not English speaking.

### INSTRUMENTATION

This study used the Pectus Carinatum Evaluation Questionnaire (Appendix A, SDC 1: http://links.lww.com/JPO/A63), a self-report outcome measure designed to measure compliance, symptoms, social influence, and engagement with daily activity during orthotic intervention for PC. This survey has evidence of validity[13] in mixed-gender pediatric populations. It is the only self-report clinical tool designed specifically for PC treatment and consists of 23 questions: 4 on compliance, 7 on adaptation, 6 on social influence, and 6 on activities. The data collected are primarily ordinal, collecting either a ranking on a scale of 0 to 10, or asking the participant to evaluate impact by responding with “always,” “many times,” “sometimes,” “few times,” or “never.” The last section requests patients respond to questions with the following answers: “not at all,” “yes, a little,” or “yes, a lot.” The PCEQ was modified slightly: instead of asking for the date of the end of treatment, we asked for the date the subject transitioned to nighttime-only wear, as this marks the end of corrective treatment. We added one question regarding satisfaction with care at the end of the questionnaire to accompany the feedback request, which was a yes/no question.

### PROCEDURE

The survey was administered primarily online. Study data were collected and managed using Research Electronic Data Capture (REDCap) electronic data capture tools hosted at

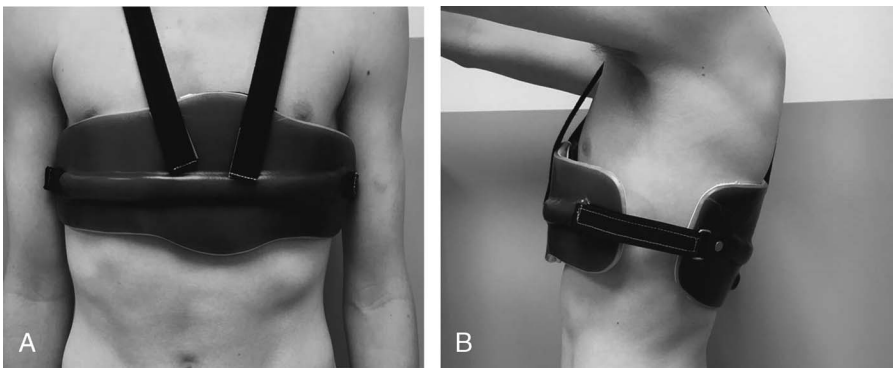


**Figure 1.** A, Anterior view of the UCSF-PCO. B, Lateral view of the UCSF-PCO. UCSF-PCO indicates custom University of California, San Francisco, Pectus Carinatum Orthosis.



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UCSF.[14,15] Research Electronic Data Capture is a secure, Web-based software platform designed to support data capture for research studies, providing 1) an intuitive interface for validated data capture; 2) audit trails for tracking data manipulation and export procedures; 3) automated export procedures for seamless data downloads to common statistical packages; and 4) procedures for data integration and interoperability with external sources. The link to the survey was emailed directly to patients, and each patient was given a unique four-digit identification number to input such that they could return and complete the survey at any time. Participants were also given the option to have the survey mailed to their homes. Reminder emails were sent bimonthly to encourage completion of the survey. Survey results were deidentified for the purposes of data analysis. Data were collected by clinicians at UCSF who did not have previous contact with these patients.

Once the subjects accessed the survey, they were prompted for signatures for subject consent and parental assent. This was required to proceed to the survey. Subjects and parents were also required to sign a Health Insurance Portability and Accountability Act authorization form. This study was granted approval by the UCSF Institutional Review Board.

### DATA ANALYSIS

The data were compiled in REDCap and summarized using nonparametric descriptive statistics. Because of the largely ordinal data set and the small sample size, parametric statistics were not appropriate.[16] The selected statistics were median and interquartile range (IQR), which give a measure of central tendency that is not heavily influenced by outliers, as well as frequency and range.

## RESULTS

### SUBJECTS

Fifty-seven patients were identified as potential subjects. Of these, 16 could not be contacted and 6 declined to participate. Of the 35 who verbally consented, 3 later withdrew and 20 did not complete the provided survey within the time frame of data collection. The remaining 12 subjects (11 male patients and 1 female patient) had an average age of 15 years (range, 12–17 years). Ten completed the survey electronically, whereas 2 returned a written copy. Of the 12 participants, 3 were still undergoing orthotic intervention; 6 subjects had ceased treatment an average of 19 months before the survey (range, 4–58 months) and had undergone orthotic treatment for an average of 16 months (range, 2–25 months). The three remaining subjects failed to provide an estimate of the date they ceased using the orthosis. See the Table for a summary of demographic data.

Table. Subject demographics (n = 12)

| Characteristics | |
|---|---|
| Age, y | |
| Mean (SD) | 15 (1.38) |
| Range | 12–17 |
| Sex, n (%) | |
| Male | 11 (92) |
| Female | 1 (8) |
| State of treatment | |
| Completed, n (%) | 6 (50) |
| Time since completion, mean, mo | 19 |
| Range, mo | 4–58 |
| Duration of treatment, mean, mo | 16 |
| Range, mo | 2–25 |
| Ongoing, n (%) | 3 (25) |
| Not reported, n (%) | 3 (25) |

### COMPLIANCE

The 12 respondents reported wearing the PCO for a median of 11 hours (range, 4–24; IQR, 16.125) daily, 5.5 days per week (range, 3–7; IQR, 3.75). Four subjects reported wearing the orthosis the full number of hours and days prescribed.

### ADAPTATION

The following negative symptoms were reported: 3 of the 12 experienced chest pain, 2 had difficulty breathing, and 1 had back pain. For subjects reporting pain, the average numerical pain score for chest pain over the last 6 months was 3.7/10 (range, 3–5), and the back pain was 3/10 in severity. Seven reported no symptoms. Five reported never being able to wear the orthosis for the full duration.

### SOCIAL INFLUENCE

Moderate levels of social influence and lower parental supervision of orthosis use were reported (Figure 2). Seven patients reported parents "many times" or "always" being strict with school duties, whereas only four reported parents being strict with wearing the orthosis. Eight subjects reported "always" or "many times" feeling uncomfortable about being seen by their friends. However, only three reported their friends making fun of them at that same frequency. Five avoided situations "many times" or "always" so that no one noticed the orthosis underneath their clothing.

### ACTIVITIES

Six respondents felt that the orthosis severely or moderately limited them in vigorous activities. No respondents felt severely limited in moderate activities, but one felt severely limited in social activities and leaning, kneeling, or bending down (Figure 3).

### FEEDBACK

Three subjects reported that the orthosis was uncomfortable, one suggested starting orthotic treatment at a younger age, one complained of skin irritation, and one requested more instruction on how to properly clean and care for the orthosis. The three subjects who reported discomfort did not report pain earlier in the survey. Eight reported they were happy with the results of their orthotic treatment.

## DISCUSSION

The results did not support the hypothesis that patients would be compliant with the UCSF-PCO. Low compliance to the Calgary protocol of 23 hours, 7 days per week was observed in number of hours more than number of days worn, although the reported wear time in this cohort was similar to the



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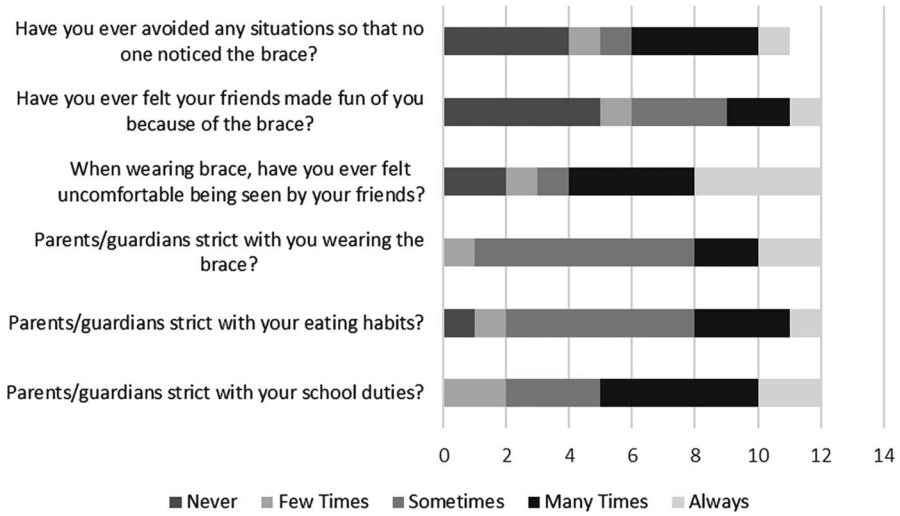


Figure 2. Social influence.

12 hours, 6 days per week of wear reported by Pessanha et al.[13] Self-reported wear time can be an unreliable measure because it requires the patient to recall previous actions with a level of accuracy that may be difficult to achieve.[17] The results did support the second hypothesis, that patients would report overall satisfaction with treatment and minimal social and physical restriction. Although patients reported moderate self-consciousness on average, only 9.1% felt severely limited in social activities. Moderate- and low-level activity limitation was reported to be minimal.

Compared with previous findings, fewer negative symptoms were associated with orthotic treatment using the UCSF-PCO. Kang et al.[9] found that pain was a factor in noncompliance with a prefabricated orthosis, with 62.8% of patients reporting pain with treatment regardless of compliance. Jung et al.[6] found that 11% of subjects treated with prefabricated orthoses discontinued treatment because of severe pain. This study had pain reported as a symptom in 25% of patients, with an average severity of 3.6/10, although the PCEQ does not specify if the pain is attributed to the orthosis or if it was present before orthotic intervention. The occurrence of pain did not seem to be linked to compliance in our study.

Orthoses remain the primary conservative method for treatment of PC. Successful orthotic treatment has been associated with reported improvements in self-esteem and increases in social activities.[3] The satisfaction rate of 72.7% is within the range of 65% to 88.4% success/satisfaction rates reported in other studies,[1,6,7,18] although it is important to note that these other studies commonly used an ordinal scale of 1 to 4 for satisfaction rates whereas the PCEQ uses a yes/no question.

Self-report feedback on patient care is beneficial in that it provides a framework for patient education and may indicate what adjustments are necessary to improve the design of the orthotic intervention. The feedback provided during this study could indicate that a more thorough discussion of expectations for the orthosis as well as how to care for it could be beneficial in reaching higher compliance rates.

Limitations of this study include the small number of respondents, sampling style, and timing of survey. With a 21% response rate and a small sample size, it is not possible to assume normal distribution of data points or that the sample is representative of the UCSF population. Patients from low socioeconomic

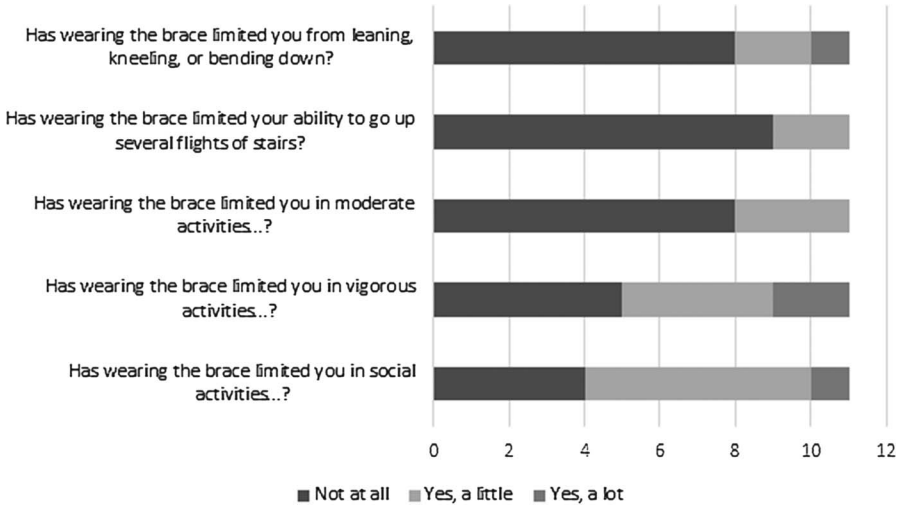


Figure 3. Activities.



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status were likely inadvertently excluded, as the treatment is not covered by all insurances. In addition, with a convenience sample, there is a high risk of bias. Respondents who completed the survey may have stronger feelings about their treatment, positive or negative. The sample is unlikely to be representative of the population of individuals who undergo orthotic treatment for PC. There is also geographic bias: subjects were patients in the San Francisco area population self-selected to people able to access care at UCSF, a tertiary referral center. As the subjects surveyed sought orthotic treatment from 2012 to 2018, it is possible for several years to have passed since the patient received the orthosis. Therefore, the report of their experience or of their wear time may not be as accurate or as reliable as if the survey had been given during or at the cessation of treatment. The survey itself had been modified slightly for this study and was administered both electronically and by paper, which may impact its psychometric properties. In addition, objective orthotic success could not be determined owing to the self-report method of data collection. Researchers participating in this study are also the clinicians using UCSF-PCO, and so they may introduce bias.

Further research is necessary to determine if the custom UCSF-PCO design reduces discomfort and leads to improved orthotic outcomes compared with the prefabricated ratchet-style orthoses commonly used. Prospective studies could address many of the limitations of this study by incorporating monitors to track wear time, recording objective reduction of the prominences, and/or randomizing patients to custom or prefabricated orthoses. Studies performed at other institutions would improve population diversity. Orthotic treatment for scoliosis has been studied extensively and has led to insurance coverage for this intervention. Further understanding of the impact of orthotic treatment for PC may lead to better insurance coverage of custom pectus orthoses.

## CONCLUSION

This study shows the potential of improved comfort and decreased activity limitations for children seeking orthotic treatment of PC as compared with designs used in previous research. However, average wear time was within the reported range of other studies, implying that there may be other factors affecting overall wear time. Based on comments provided by participants, this may include additional patient education and emphasis on the importance of wear time to full correction.

## ACKNOWLEDGMENTS

We thank Farahnas Doost, MPO, and Jeanny Ledda, MPO, for assistance with subject recruitment and data collection, as well as Sarit Cliffer, MPO, Scott Annis, CPO, and Maria Lesieutre, MPO, for invaluable feedback on the rough draft of the manuscript.

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